Redefining Knee Balance in a Medially Stabilized Prosthesis: An In-Vitro Study

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Abstract: Background To date, there is still no consensus on what soft tissues must be preserved and what structures can be safely released during total knee arthroplasty (TKA) with a medially stabilized implant. Objective The aim of this study was to analyze the effect of a progressive selective release of the medial and lateral soft tissues in a knee implanted with a medially stabilized prosthesis. Method Six cadaveric fresh-frozen full leg specimens were tested. In each case, kinematic pattern and mediolateral laxity were measured in three stages: firstly, prior to implantation; secondly, after the implantation of the trial components, but before any soft tissue release; and thirdly, progressively as soft tissue was released with the trial implant in place. The incremental impact of each selective release on knee balance was then analyzed. Results In all cases sagittal stability was not affected by the progressive release of the lateral soft tissue envelope. It was possible to perform progressive lateral release provided the anterior one-third of the iliotibial band (ITB) remained intact. Progressive medial release could be performed on the medial side provided the anterior fibers of the superficial medial collateral ligament (sMCL) remained intact. Conclusion The medially conforming implant remains stable provided the anterior fibers of sMCL and the anterior fibers of the ITB remain intact. The implant's sagittal stability is mainly dependent on its medial ball-in-socket design.

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ORIGINAL ARTICLE

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Mid- to long-term results of total ankle replacement in patients with haemophilic arthropathy: A 10-year follow-up

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Introduction: Haemophilic ankle arthropathy is caused by recurrent spontaneous joint haemorrhaging and leads to pain, deformity and loss of function. In the presence of advanced articular deterioration, therapeutic options are confined to either arthroplasty or arthrodesis, the latter still being referred to as the procedure of choice. However, total ankle replacement (TAR) has recently gained acceptance as an alternative.

Aim: To investigate the mid- to long-term results of TAR in haemophilic ankle arthropathy.

Materials and Methods: Seventeen TARs in 14 male patients (mean age: 43 years [range, 27.4-57.6]), implanted between 1998 and 2012, were retrospectively analysed. Implant survival was estimated using Kaplan-Meier analysis. Haemophilic/viral status, complications and revision surgeries were recorded. Follow-up assessment of 12 TARs was performed 9.6 years (range, 3.3-17.8) postoperatively, including clinical examination, pain and satisfaction scales, the American Orthopaedic Foot and Ankle Society (AOFAS) hindfoot score, and the SF-36. Radiographic evaluation of pre- and follow-up radiographs was conducted.

Results: Estimated implant survival was 94% at 5, 85% at 10 and 70% at 15 years, respectively. Three cases required revision surgery. At follow-up, 9.6 years (range, 3.3-17.8) postoperatively, the level of satisfaction was 76% (range, 50-100) and of pain 2/10 (range, 0-6) on the VAS. Range of motion had increased significantly (P = .037). The SF-36 summary scores were comparable to those of a matched standard population. The AOFAS hindfoot score averaged 81 points (range, 73-90). All radiographs revealed component loosening or periprosthetic radiolucency.

Conclusion: Total ankle replacement in the presence of advanced haemophilic arthropathy is a viable treatment option with favourable mid-/long-term results, maintaining mobility of the ankle joint.

KEYWORDS

hemophilia, hemophilic arthropathy, implant survival, total ankle replacement

1 | INTRODUCTION

Haemophilia is a rare, congenital, haematological disease (approximately 1 in 30 000 live male births[1]) associated with recurrent spontaneous haemorrhaging. Approximately 70% of all bleeding events involve the joints,[2] eventually causing haemophilic arthropathy. Knees elbows and ankles are most commonly affected. Symptoms comprise joint pain and deformity, paired with loss of function. While haemophilia A (lack of factor VIII) and B (lack of factor IX) are X-linked hereditary disorders only affecting male subjects, other factor deficiencies may result in the same cycle of progressive destruction of target joints. Affected persons usually require lifelong medical attention and intricate treatment plans provided by a multidisciplinary team of specialized health care professionals. Early diagnosis, patient training and monitoring, as well as rigorous substitution of the deficient clotting factor, are of paramount importance to delay adverse effects such as haemophilic arthropathy.

Once severe joint deterioration has occurred, and symptoms are no longer manageable by means of non-operative treatment or joint preserving procedures, further therapeutic options are confined to either prosthetic joint replacement or arthrodesis. With regard to the ankle joint, current literature still refers to arthrodesis as the procedure of choice,[3,4] offering pain relief and favourable functional outcomes, with minimal rates of postoperative complications.[5] However, more recently total ankle replacement (TAR) has gained acceptance as an alternative in this setting. An increasing number of publications provide evidence of its capacity to reduce pain significantly, while preserving ankle motion, thereby potentially reducing adjacent joint disease.[6-8] As the development and successes of TAR for said condition are of recent nature, there is still a paucity of long-term outcome reports. The objective of this study was, therefore, to investigate the mid- to long-term clinical and radiographic results after TAR in patients with haemophilic ankle arthropathy.

2 | MATERIALS AND METHODS

2.1 | Patients

For the purpose of this observational study, we searched our institutional database for all cases of haemophilic patients who underwent primary TAR between 1998 and 2012. Seventeen consecutive cases in 14 male patients (age: 43 [range, 27.4-57.6 years]) were identified. All patients provided informed consent. Approval of the responsible ethics committee (Kantonale Ethikkommission Zürich) was obtained. To facilitate readability of this article, we will in the following refer to cases rather than patients.

As illustrated in Figure 1, pre- and perioperative demographic and disease-specific data, as well as implant survival, were analysed in all 17 cases. Two cases were not available for a follow-up visit. However, revision-free implant survival was confirmed via telephone interview. Three ankles had undergone revision surgery, two of which revision TAR (rTAR) and one arthrodesis. Thus, with a mean follow-up of 9.6 (range, 3.3-17.8), 12 cases were seen for follow-up examination.

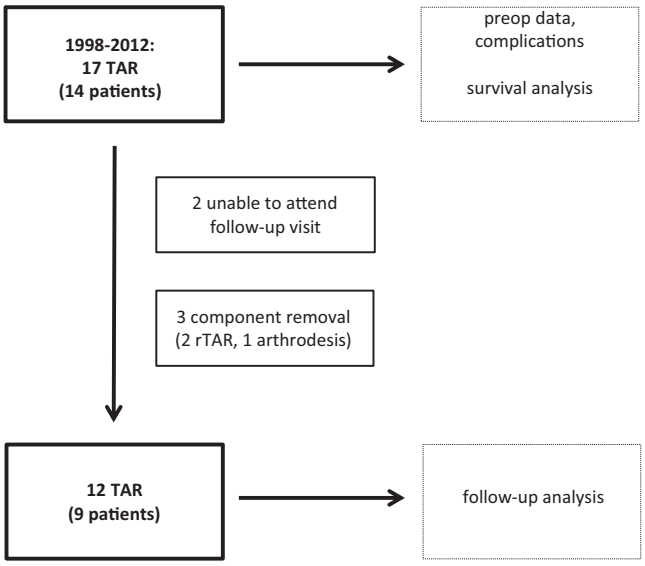

FIGURE 1 Patient/case selection and analyses

An overview of the study population, basic demographics and disease-specific data are presented in Table 1. Eleven patients (78.6%) were diagnosed with haemophilia A, 2 patients (14.3%) with haemophilia B and one patient (7.1%) with a congenital factor VII deficiency. Thirteen surgeries (76.5) were indicated in the setting of severe, 3 (17.6%) in the setting of moderate and one (5.9%) in the setting of mild haemophilia† .[9] One patient was HIV positive (6.9%) and 13 were diagnosed with chronic hepatitis B and/or C prior to surgery (76.5%). The mean preoperative Pettersson score was 7.8 ± 6.8 (range, 5-10).

2.2 | Surgical intervention

During the course of the 15-year long observation period, 4 different implants were used, 6 AGILITY (DePuy, Leeds, United Kingdom), 6 HINTEGRA (Newdeal SA, Lyon, France), 3 STAR (Link, Hamburg, Germany) and 2 MOBILITY (DePuy). All surgeries were performed by the institution's senior foot and ankle surgeon at the time (PV, NE, JS). In 11 cases, surgical lengthening of the Achilles tendon/Gastrocnemius was carried out to accomplish a minimum of 10° of intraoperative ankle dorsiflexion. Once, an additional talonavicular arthrodesis, thrice, additional subtalar arthrodeses were necessary due to concomitant arthropathy of the respective joints. In one case, supplemental medializing calcaneal osteotomy for correction of hindfoot alignment was carried out.

2.3 | Peri-/postoperative treatment

Perioperative factor replacement therapy was overseen by a senior consultant haematologist. Both a careful review of recent bleeding history and laboratory analyses including measurement of clotting

†Classification complies with the International Society on Thrombosis and Haemostasis (ISTH) standard and is based on clotting factor activity: severe <1%, moderate 1%-5%, mild 6%-40%.

TABLE 1 Overview: demographic and disease-specific data of 17 total ankle replacements (TAR) in haemophilic patients

	Absolute (n/17)	Proportion (%)	Mean	SD	Min	Max
Unable to attend follow-up visit	2	11.8				
Revisions with component removal[a]	3	17.6				
Time to revision (y)			7.5	4.9	2.2	10.8
Prosthetic survival	14	82.4				
Follow-up (y)			9.3	4.9	2.2	17.8
Prosthetic survival and follow-up visit	12	70.6				
Follow-up (y)			9.6	5.4	3.3	17.8
Gender	All male					
Age at surgery (y)			43.4	11.1	27.4	57.6
BMI (kg/m²)			24.7	4	17.7	30.4
Smokers	4	23.5				
Haemophilia A	11	78.6				
Haemophilia B	2	14.3				
Factor VII deficiency	1	7.1				
Severe haemophilia	13	76.5				
Moderate haemophilia	3	17.6				
Mild haemophilia	1	5.9				
Factor substitution on demand	12	85.7				
Factor substitution regularly	2	14.3				
HIV positive	1	6.9				
Chronic hepatitis B and/or C	13	76.5				
Pettersson score			7.8	6.8	5	10

[a]1 × arthrodesis, 2 × revision TAR.

factor activity and antibody screening were obtained. Based on these preoperative parameters, repeat measurements of factor activity, intraoperative blood loss and postoperative bleeding tendency, individual treatment schemes for intravenous factor substitution were implemented. A preoperative loading dose aiming for 100% factor activity was followed by bolus injections in decreasing frequency, keeping factor activity between 70% and 90% during the first 3 days, and above 50% until day 8. Patients were released from inpatient treatment only when systemic and local control of the haemorrhagic diathesis was obtained. The duration of this early postoperative phase necessitating close coagulation management corresponds to the duration of hospitalization which counted 14.6 ± 4.3 days (range, 8-26).

All patients received basic postoperative wound and pain management. Those without additional osseous interventions were allowed to start full weight bearing in a cast or VACO®ped (Oped AG, Cham, Switzerland) after few days of bed rest. As soon as superficial wound healing was achieved, passive ankle mobilization was started to increase range of motion (ROM). Those with additional osseous procedures were immobilized in a non-weight-bearing cast for 6 weeks. All patients transitioned to full weight-bearing not later than 3 months postoperatively, depending on the healing process and level of pain.

2.4 | Clinical assessment

At the follow-up visit, patients underwent an interview and a detailed physical examination conducted by a senior resident (FE) not involved in the surgical treatment. Subjective levels of satisfaction (0%-100%) and pain (0-10, visual analogue scale [VAS]) were recorded. Postoperative ROM was measured with a goniometer along the lateral boarder of the leg and foot in suspension, and compared to preoperative ROM, which could be extracted from the patients' files. The American Orthopaedic Foot and Ankle Society (AOFAS) hindfoot score[10,11] and the SF-36 (Medical Outcome Study 36-Item Short-Form Health Survey)[12] were obtained. The sample group's SF-36 summery scores for physical and mental health were calculated based on those of a gender- and

age-matched reference group (German norm population 1998, male, age 51-60).[13] Patients' ability to participate in working life and activities of daily living (ADL), as well as their need for orthopaedic aids (yes/no, type), was inquired.

2.5 | Radiographic assessment

All radiographs were obtained in a weight-bearing position. Radiological evaluation was performed by a board-certified radiologist specialized in musculoskeletal radiology. The preoperative severity of haemophilic arthropathy was graded employing the Pettersson score.[14] As proposed by Lecomte[15] and adopted by Mulcahy et al,[16] postoperative radiographs were examined regarding component position, component loosening (subsidence, migration, angulation), periprosthetic lucency, periprosthetic fracture, polyethylene wear or fracture, soft tissue abnormality and progressive degenerative changes in adjacent joints. Component position was assessed on standard anteroposterior ankle radiographs by measuring the angle between the midline of the tibial and the talar component. Angles between 3° varus and 3° valgus were considered unremarkable. Ankle positions exceeding this range were considered mal-aligned as they may result in component edge-loading/-wear, impingement, and/or excessive unilateral foot pressure.[15,17,18]

2.6 | Statistical analysis

Statistical analysis was performed with IBM SPSS Statistics, Version 21.0 (IBM Corp., Armonk, NY). Data are presented as mean, standard deviation (SD) and range for continuous variables and as proportion (%) for categorical variables. The Kolmogorov-Smirnov test was applied to test for normality of distribution of data. Kaplan-Meier analysis with 95% confidence intervals (CI) was performed to estimate prosthetic survival. The Wilcoxon signed-rank test was used for testing differences between means. A *P*-value of <.05 was considered to be statistically significant.

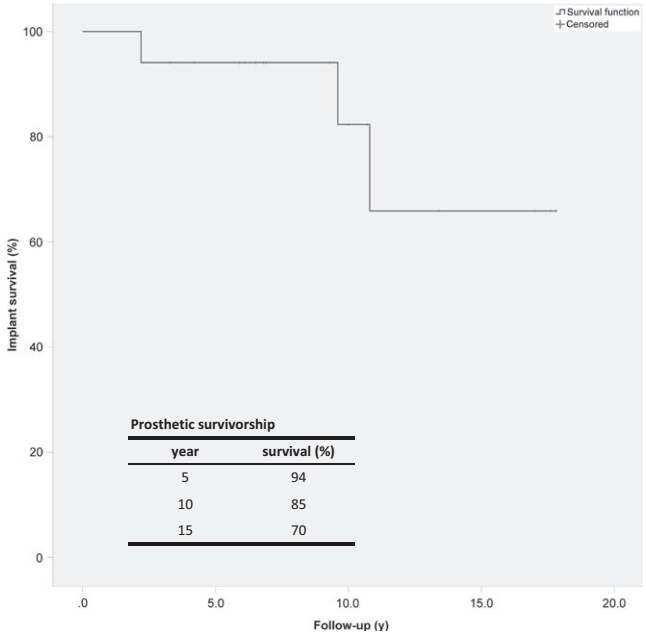

FIGURE 2 Estimated implant survival based on Kaplan-Meier analysis

3 | RESULTS

3.1 | Implant survival

Implant survival was assessed for all 17 cases after a mean follow-up of 9.3 years (range, 2.2-17.8). As a result of component loosening, 3 cases (17.6%) had undergone revision surgery with component removal (one arthrodesis, 2 rTAR) at a mean postoperative interval of 7.5 ± 4.9 years (range, 2.2-10.8). Based on Kaplan-Meier curves, the estimated implant survival was 94% at 5, 85% at 10 and 70% at 15 years. The mean estimated implant survival was 14.77 years (95% CI, 11.9-17.7; Figure 2).

3.2 | Postoperative clinical outcome

A comprehensive synopsis of all clinical outcome parameters is offered in Table 2. The levels of satisfaction and pain were 76 ± 11.8% (range, 50-100) and 1.9/10 ± 2.0 (range, 0-6) on the VAS, respectively. An increase in ROM was observed in 10 cases (83%) and a decrease in 2 cases (17%). Overall, ROM increased significantly (*P* = .037) by an average of 10.2 ± 16.5° (range, −30 to 32). The AOFAS hindfoot score reached 81 ± 5.9 points (range, 73-90). Regarding the SF-36, the study population's summery scores were 47 ± 13.0 (range, 16-59) for physical and 57 ± 5.0 (range, 50-65) for mental health. The corresponding values for an age- and gender-matched norm population are 47 and 52, respectively [19] (Table 3). All patients were able to pursue work and activities of daily living (ADL). Stair climbing was reported to be associated with no difficulties in one case, with minor difficulties in 10 and major difficulties in one case. Engaging in sport activities was hardly possible. Except for orthopaedic footwear (3 × custom-made orthopaedic shoes; 1 × standard shoes with adjustments), no orthopaedic aids were needed.

3.3 | Radiographic assessment

A detailed case by case evaluation of all 12 ankles is presented in Table 4. Six prostheses were considered well-aligned (5 × 2° valgus; 0°) and 6 mal-aligned (4, 5, 10, 12, 13° valgus; 4° varus). All radiographs displayed signs of component loosening: Subsidence was observed 5 times on the tibial side and 6 times on the talar side, component migration once on the tibial and 4 times on the talar side and angulation twice on the talar side. Periprosthetic radiolucency was present in 9 cases tibially and in 11 cases talarly. There were no periprosthetic fractures. Component wear or fracture as well as adjacent degenerative joint disease was rare. Subtalar degeneration was found in 3 cases. On the level of Chopart's joint line, no degenerative changes were identified.

TABLE 2 Synopsis of individual clinical follow-up data of 12 cases of total ankle replacement in haemophilic patients with a follow-up of 9.6 y

Case	Follow-up (y)	Age (y)	Satisfaction (max. 100%)	VAS (max. 10)	ROM (°)				AOFAS (max. 100)	SF-36 summary scores[a]	
					Pre	Post	Δ ROM	Gain (↑) loss (↓) n/12 [%]		Physical health	Mental health
1	6.8	54	75	6	25	20	−5	↓	73	47	50
2	4.2	60	70	0	10	25	15	↑	89	48	51
3	5.9	60	70	0	10	15	5	↑	89	48	51
4	13.4	62	75	3	5	15	10	↑	76	59	65
5	6.5	34	80	2	10	30	20	↑	80	56	60
6	6.9	34	80	2	20	30	10	↑	80	56	60
7	10.0	61	90	2	15	35	20	↑	76	16	57
8	17.0	53	60	0	5	10	5	↑	87	39	58
9	17.6	54	100	0	8	40	32	↑	83	49	59
10	17.8	54	100	0	15	45	30	↑	73	49	59
11	3.3	34	50	5	45	15	−30	↓	79	44	63
12	6.1	54	65	3	25	35	10	↑	90	49	56
Mean	9.6	51.1	76	1.9	16	26	10.2	10/12 (83) ↑	81	47	57
SD	5.4	11.0	11.8	2.0	11.4	11.3	16.5		6.3	11.1	4.7
Min	3.3	34	50	0	5	10	−30	2/12 (17) ↓	73	16	50
Max	17.8	62	100	6	45	45	32		90	59	65
							$P = .037$[b]				

Working/able to pursue activities of daily living	Difficulties stairs	Difficulties sports	Orthopaedic aids
Yes	Minor	Major	None
Yes	Minor	Major	Custom-made shoes
Yes	Minor	Major	Custom-made shoes
Yes	Minor	Impossible	None
Yes	Minor	Major	None
Yes	Minor	Major	None
Yes	Major	Impossible	Custom-made shoes
Yes	None	Impossible	None
Yes	Minor	Minor	None
Yes	Minor	Minor	None
Yes	Minor	Major	Adjusted shoes
Yes	Minor	Minor	None

AOFAS, American Orthopaedic Foot and Ankle Society; ROM, range of motion.

[a]On a scale from 0 to 100 points, the scores transmit the respondent's subjective level of disability. 0 points correspond to full disability, 100 points to no disability.

[b]Wilcoxon signed-rank test.

TABLE 3 SF-36: study population vs standard population[a]

	Study population (male, Ø 51 y)	Standard population (male, 51-60 y)
Summary score physical health	47	47
Summary score mental health	57	52

[a]German norm population 1998, male, age 51-60.[12]

3.4 | Complications

Peri- and postoperative complications are listed in Table 5. Two intraoperative fractures, one of the lateral and one of the medial malleolus occurred, were immediately addressed with osteosynthesis. Neither the evaluation of the 2 patients' files nor the assessment of their imagery was indicative of problems with fracture healing. However, both

TABLE 4 Synopsis of individual radiographic follow-up of 12 cases of TAR in haemophilic patients with a follow-up of 9.6 y

Case	Follow-up (y)	Position	Component loosening		Periprosthetic lucency		Soft tissue abnormality	Periprosthetic fracture	Component wear/fracture	Degeneration	
			Tibia	Talus	Tibia	Talus				Subtalar	Chopart
1	6.8	4° valgus	None	Subsidence	Yes	Yes	HO	No	No	Fused	No
2	4.2	5° valgus	None	Subsidence	Yes	Yes	HO	No	No	Yes	No
3	5.9	4° varus	None	Migration	Yes	Yes	HO	No	Yes	No	No
4	13.4	10° valgus	Subsidence	Angulation	Yes	Yes	Swelling	No	No	Fused	Fused
5	6.5	12° valgus	Subsidence	Subsidence	Yes	Yes	Swelling	No	No	No	No
6	6.9	2° valgus	None	Migration	Yes	Yes	HO	No	No	No	No
7	10.0	2° valgus	None	Subsidence	No	No	HO	No	No	No	No
8	17.0	2° valgus	Subsidence	Migration	Yes	Yes	HO	No	No	No	No
9	17.6	2° valgus	Subsidence	Subsidence	Yes	Yes	HO	No	No	Yes	No
10	17.8	2° valgus	Migration	Subsidence	No	Yes	HO	No	No	Yes	No
11	3.3	0°	None	Migration	No	Yes	HO	No	No	No	No
12	6.1	13° valgus	Subsidence	Angulation	Yes	Yes	HO	No	No	No	No

Ho, hypertrophic ossification.

patients were later revised secondary to prosthetic loosening, suggesting a correlation between the intraoperative and the late complication. The patient with the medial malleolar fracture also sustained a lesion of the medial plantar nerve resulting in hypoesthesia and neuropathic pain which was treated with benzodiazepines and pregabalin. One postoperative haematoma spontaneously resolved during a prolonged hospital stay of 22 days. None of the patients developed wound-healing disorders, bleeding or infectious complications. In one case, a calcaneal and a first metatarsal stress fracture were diagnosed at the 4-month follow-up visit following onset of pain 2 weeks earlier. As symptoms had already subsided and the patient was unwilling to follow restrictions, no treatment was implemented. Two ankles underwent soft tissue revision to improve ROM in the presence of symptomatic, reduced dorsiflexion. Three ankles underwent revision surgery with component removal, two of which rTAR and one arthrodesis. Both rTAR were performed due to symptomatic loosening of the talar component. In one case, only the talar component was exchanged (HINTEGRA Revision Ankle Prosthesis [Newdeal SA]), in the other the TAR was entirely replaced (SALTO TALARIS XT Revision TAR [Tornier, Inc., Bloomington, MN, USA]). The conversion to ankle fusion was achieved by anterior approach and plate osteosynthesis (INTEGRA TIBIAXYS Ankle Fusion [Newdeal SA]) with interposition of both autologous cancellous bone harvested from the iliac crest and allograft.

4 | DISCUSSION

Haemophilic arthropathy is caused by recurrent spontaneous haemorrhaging and leads to articular deterioration. If symptoms are no longer manageable by means of non-operative treatment or joint preserving procedures, arthrodesis or prosthetic joint replacement needs to be considered. While arthrodesis has been known as the treatment of choice,[20] TAR has recently been demonstrated to be a viable alternative with favourable short- and midterm outcome.[6,7,21,22] However, data documenting the long-term outcome are sparse. Therefore, the aim of this study was to report the mid- to long-term results of TAR in patients with haemophilic arthropathy.

The estimated implant survival in this case series was 94% at 5, 85% at 10 and 70% at 15 years. Corresponding studies reporting long-term outcomes in haemophiliacs are missing. However, the here reported long-term survival is comparable both to the results in patients with inflammatory joint disease [23] and to the results in a general population. As haemophilic arthropathy and inflammatory joint disease share some pathophysiological aspects,[24] comparison seems particularly applicable. In the light of this, the present study's 15-year implant survival of 70% nearly accords with the 73% reported by Kraal et al[23] who examined 93 cases of TAR in inflammatory joint disease. Bartel et al[25] reported 5- and 10-year survival rates of 87% and 81% (vs 94% and 85%), based on data of all 6 listed national joint registries including 5152 primary TARs in general populations. Yet, 2 characteristics of haemophilic patients set them apart. Firstly, microhaemorrhages around prosthetic components may cause destructive reactions leading to increased rates of aseptic loosening.[26] An example of such loosening is

TABLE 5 Peri-/postoperative complications

Number	Type	Management	Comment
2	Intraoperative fracture (1 × lateral, 1 × medial malleolus)	Osteosynthesis	Both patients later underwent removal of TAR
1	Nerve lesion (medial plantar nerve)	Medical treatment of neuropathic pain	Same patient suffered intraoperative fracture of medial malleolus
1	Haematoma	Non-operative treatment	
1	Stress fracture (calcaneus and first metatarsal)	None	Diagnosed retrospectively
2	Unsatisfying ROM, reduced dorsiflexion	Soft tissue revision	Both 11 mo postoperatively
3	Component loosening, associated with mechanical pain	Removal of TAR	2 rTAR (2.2/9.6 y postop), 1 arthrodesis 10.8 y postop)

ROM, range of motion; TAR, total ankle replacement.

illustrated in Figure 3. Secondly, patient age at time of surgery tends to be younger than in other populations leading to assumedly higher demands. It is still a matter of discussion though, to which degree patient age at time of TAR implantation affects prosthetic longevity.[27-29]

The clinical outcome of this study group was good, as represented by an AOFAS score which was situated at the upper end of the reported spectrum.[7,21,30] However, while high levels of satisfaction, little pain and few restrictions regarding ADL were reported, all follow-up radiographs showed signs of either component loosening or periprosthetic lucency. Furthermore, half of the examined TAR was rated to be mal-aligned. These paradoxical clinical and radiographic findings may be partly attributable to the concept of silent osteolysis which has been described in the context of joint replacement, rendering patients asymptomatic in spite of the presence of signs of component loosening.[31,32] Moreover and along the lines of this present study, good clinical midterm results of TAR despite mild mal-alignment have been reported before.[33]

The average gain of ROM (10°) and the mean postoperative motion arc (26°) were satisfactory except during stair climbing, an action requiring dorsiflexion, above all. Thus, the importance of reaching and preserving a minimum of 10° during implantation and rehabilitation is highlighted.[17] The question, whether or not preservation of ankle motion is beneficial in the long run, and when compared to fusion, remains unclear though. Remarkably, Stavrakis[34] found that between 8491 arthrodeses and 1280 ankle replacements, patients with TAR were less likely to need subsequent subtalar arthrodesis. The absence of symptomatic adjacent joint disease within our study group supports this hypothesis.

In discordance with previous publications using the SF-36 for measurement of health-related quality of life in patients with coagulation disorders,[35,36] the perception of physical health in this population was equal to that of a designated reference group. Mental health was graded even superiorly. We hypothesize that the high level of subjective satisfaction stems from the comprehensive multidisciplinary care these patients receive from early on.

The incidence of mechanical complications (intraoperative/stress fracture, nerve lesion, unsatisfying ROM, component loosening) corresponded to the literature.[37,38] Malleolar fractures, for example, occur in up to 10% of during TAR implantation.[39,40] Due to poor subchondral bone quality, the risk in haemophilic patients may be particularly high.

Notably, none of the patients developed bleeding complications. Furthermore, no revision surgeries for treatment of wound or periprosthetic joint infection were necessary, although the diagnosis of haemophilia with concomitant hepatitides constitutes a known risk factor [41] and although the infection rate after TAR is documented to be 2%-8.6%.[42] Only few other studies discussing septic complications after TAR in haemophilic patients can are available for comparison. Strauss et al,[21] with a series of 11 cases, reported 2 implant removals due to infection. Barg et al[7] observed no infections in a series of 10.

This study has limitations. Firstly, this is a retrospective study. Also, due to incomplete preoperative scores, we were not able to longitudinally analyse all outcome parameters thereby missing out on potentially meaningful information. Secondly, while the majority of the study collective experienced an increase in ROM, we are aware that this gain may be related mostly to the reduction in pain and not necessarily to a mechanical advantage. Further concerning this matter, measurements were performed clinically using a goniometer, allowing for a certain inaccuracy due to both error in measurement and the unquantifiable degree of midfoot motion in combination with tibiotalar motion.[43] Radiographic measurements were not performed. Thirdly, the AOFAS ankle/hindfoot score is a non-validated score.[44] Despite these limitations, the presented results provide the longest follow-up of TAR in haemophilic patients, thereby expanding the scope of data and knowledge on this subject relevantly and serving future investigations as reference.

5 | CONCLUSION

Total ankle replacement in the setting of advanced haemophilic ankle arthropathy is a viable treatment option with favourable clinical mid-/long-term results. Implant survival based on a 10-year follow-up is similar as in inflammatory joint disease and general populations. Preservation of ankle mobility appears to be advantageous in regard to patient comfort and occurrence of symptomatic adjacent joint disease. While clinical results are encouraging, follow-up radiographs reveal the presence of component loosening and periprosthetic radiolucency in the majority of cases. Bearing in mind the patient population's young age and specific risk factors, the need for revision surgery

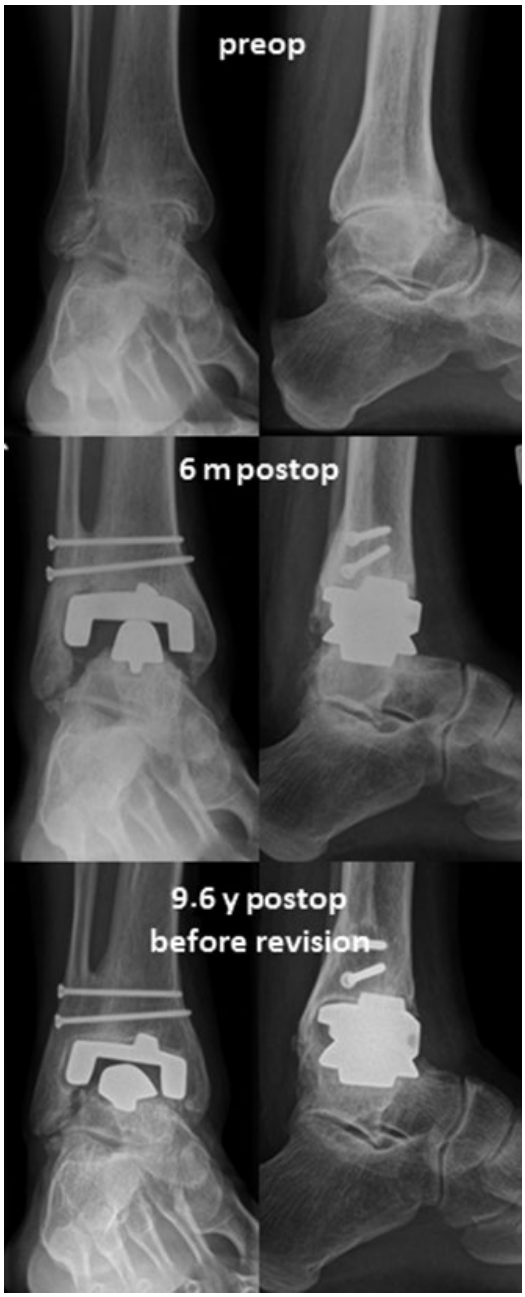

FIGURE 3 Serial radiographs of a patient with revision secondary to implant loosening 9.6 y after total ankle replacement implantation

resulting from symptomatic component loosening may arise in the long run. Further studies including a larger number of cases and even longer follow-up periods are warranted.

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DISCLOSURES

The authors stated that they had no interests which might be perceived as posing a conflict or bias.

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